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Case Report

Imaging modalities in early diagnosis of splenic ectopic pregnancy: A case report

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Abstract
Splenic ectopic pregnancy is extremely rare but carries a high risk of life-threatening intraperitoneal bleeding. Here, we present a 40-year-old woman presenting with vaginal bleeding. Although the intrauterine device (IUD) had been in place for 6 years, urinary and serum tests were positive for pregnancy. However, transvaginal ultrasound showed an empty uterus, no apparent adnexal masses or free fluid. An abdominal ultrasound was subsequently performed, which revealed a viable gestational sac in the spleen. Magnetic resonance imaging (MRI) that did not induce ionizing radiation was also performed, confirming the diagnosis of this splenic pregnancy. The gastrointestinal surgeon completed a laparotomy, which successfully removed the spleen's superior pole containing an ectopic pregnancy.

Keywords
Splenic Pregnancy, Ectopic Pregnancy, Ultrasound, MRI, Partial Splenectomy

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Introduction

The most common site of ectopic implantation is within the fallopian tube, accounting for 95.5% of all ectopic pregnancies, the remaining extra-tubal ectopic pregnancies include ovaries and abdomen, accounting for 3.2% and 1.3%, respectively [1]. Abdominal pregnancies in various extra-pelvic organs have been described, with the spleen being one of the rarest sites. A systematic review of the literature by Poole A et al. showed that in 225 abdominal ectopic pregnancies, splenic gestations accounted for only 5.3%. The remaining sites were divided into the following categories in descending order of the number of reported cases: pouches around uterus (24.4%), uterus-adnexa (24.0%), multiple abdominal organs (12.9%), omental (11.1%), bowel-appendix (6.7%), hepatic (5.8%), retroperitoneal (4.5%), abdominal wall (3.1%) and others (2.2%) [2].

Primary splenic pregnancy was common at a young maternal age (27.3-28.7), with few births (the average parity was 1.1) [2]. As with other abdominal pregnancy sites, splenic pregnancy occurred with several risk factors, including a history of pelvic inflammatory disease, endometriosis, in vitro fertilization (IVF), previous pelvic surgery, previous ectopic pregnancy, utero-tubal anomalies, and intrauterine device (IUD) [3,4]. Most cases of symptomatic splenic pregnancy are diagnosed between the 6th and 8th weeks of gestation and tend to occur earlier than in other sites of the abdomen [2,5,6], but later than tubal ectopic pregnancy [1]. Kalof et al. postulated that most splenic pregnancies at clinical presentation are 2.0 and 3.5 cm in size, and suggested that the risk of rupture in ectopic gestation exceeds this size [6].

Herein, we would like to present a case, preoperatively diagnosed by abdominal ultrasound and MRI as a splenic pregnancy with live embryos implanted at the superior pole of the spleen in a middle-aged woman who had an IUD in situ, successfully treated by partial splenectomy before its rupture.

Case Report

A 40-year-old woman, gravida 2 para 2, was admitted to the Obstetrics and Gynecology Department due to intermittent vaginal bleeding and vague pain in the lower abdomen for several days. The patient had a history of regular menses (28-day cycle), and her last menstruation was 6 weeks prior. She denied a history of pelvic inflammatory disease, tubal surgery or a previous ectopic pregnancy, except for a copper IUD 6 years ago.

At admission, she had neither vomiting nor fever, physical examination showed no abdominal tenderness or rigidity and a blood pressure of 110/60 mm Hg. Bimanual examination indicated a normal cervix and minimal bleeding discharge with no tenderness of the uterus or adnexa.

Urine pregnancy test showed positive results and serum beta-human chorionic gonadotropin (βHCG) showed 34,279 IU/L (normal <5.3). Blood laboratory tests were unremarkable with a normal red blood cell count of 4.5 T/l (4.0-5.2). However, transvaginal ultrasound revealed a normal-sized uterus with a 7.6 mm thick endometrium, but with an IUD, no identifiable intrauterine gestational sac and no signs of ectopic tubal pregnancy, normal bilateral ovaries and no free fluid in the pouch of Douglas.

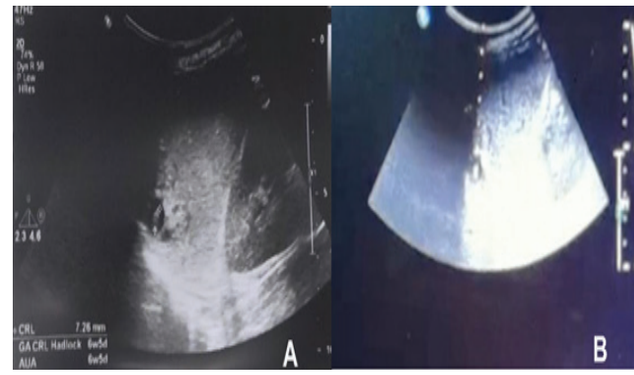

Figure 1. Abdominal ultrasound at the superior splenic pole showing a gestational sac containing the embryos (A) in the presence of a beating heart on pulsed Doppler (B).

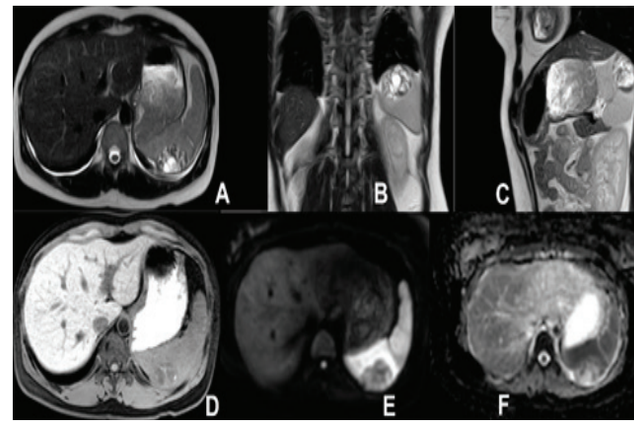

Figure 2. Abdomino-pelvic MRI. T2WI axial (A), coronal (B) and sagittal (C); T1WI VIBE FatSat (D); DWI b=800 (E) and ADC (F). The images show a mass located in the spleen adjacent to the left diaphragm, cystic component, well-margined, with many small septal in the periphery (A, B and C), interspersed with hyperintense foci of blood stasis (D) and restricted diffusion edema of perilesional splenic parenchyma (E and F).

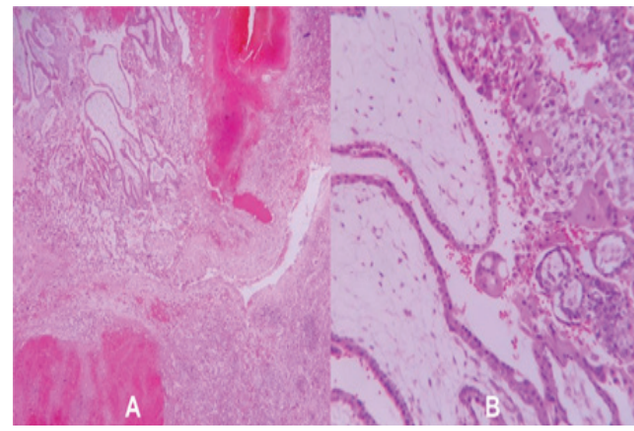

Figure 3. Histopathologic sample. A (H&E, ×50), chorionic villi invading normal splenic parenchyma, containing lymphoid follicles and adjacent areas of hemorrhage. B (H&E, ×400), chorionic villi and trophoblastic tissue.

After suspecting an extra-tubal pregnancy, further pan-abdominal ultrasound was performed to look for uncommon implantation sites. The splenic parenchyma at the superior aspect showed an ill-defined heterogeneous cystic mass measuring 36 x 47 mm, which contained an 11 mm gestational sac containing an embryo with a crown-to-rump length of 7.2 mm (equivalent to 6 weeks 4 days fetus), and fetal heart movement was confirmed at 145 beats/min (Figure 1). Abdominal ultrasound also noted the absence of free fluid in the perisplenic space.

Although an abdominal ultrasound strongly identified a splenic pregnancy, an abdominopelvic MRI was performed to obtain additional information. This technique confirmed the existence of a mixed cystic lesion measuring 28 x 37x 46 mm, accompanied by perilesional edema of the splenic parenchyma. No other abnormalities were found in the abdomen and pelvis (Figure 2). A diagnosis of primary splenic pregnancy was made. The initial surgery was an exploratory laparoscopic examination. In the upper pole of the spleen, a pregnant formation 3x4 cm in size with villous tissue, deeply embedded in the spleen parenchyma and bleeding easily, was revealed. Clearly revealing the splenic pedicle, the splenic artery branch to the upper pole is ligated, temporarily cross-clamping the inferior pole artery and manual compression, which collects blood into the vein to collapse the spleen. Subsequently, an anatomic resection of the upper pole of the spleen is performed and hemostasis with Prolene 3.0 suture. Excellent hemostasis was achieved and the abdomen was closed after washout with the a drain placed in the peri-splenic region. Postoperatively, the patient was transferred to the Intensive Care Unit for monitoring, before stepping down to the surgical ward 2 days later where the rest of her recovery was uneventful. The ß-hCG levels decreased to 908.83 IU/L 6 days after the surgery. The pelvic drain was removed on the 3rd day, and the patient was discharged on the 5th day after the operation. She had an uneventful recovery at home, and her ß-hCG levels returned to normal 4 weeks after surgery.

On sectioning in half, there was a corresponding oval mass with well-delineated but non-encapsulated dimensions of 20 x 25 x 35 mm, containing cystic and pinkish-white soft tissue mixed with dark red hemorrhages. Microscopic examination demonstrated numerous chorionic villi and intermediate trophoblasts invading the splenic parenchyma (Figure 3). No malignant cells were found.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Discussion

Due to the strong peristalsis of the intestine and the dynamics of the intraperitoneal fluid flow, it is possible to carry fertilized ovum from the cul-de-sac to different abdominal cavities. The spleen provides a smooth and flat surface of the thin capsule along with its abundant blood flow nature and accessibility in the supine position, making it a relatively sustainable site for the zygote implantation. This explains why most of the pregnant masses are implanted under the capsule, protruding outward and often beyond the border of the spleen, and their location can be anywhere from the upper pole to the lower pole and hilum [6].

Although the spleen is a favorable environment for embryonic growth, unfortunately the spleen parenchyma cannot distend to support the blastocyst growth and cannot accommodate placental attachment. Therefore, splenic pregnancy is rarely detected at the end of the first trimester [5]. In complicated cases, patients complain of acute severe abdominal cramping or typically present with left upper abdominal pain radiating to the left shoulder, followed by signs of peritonitis and unstable hemodynamic status leading to urgent splenectomy [2,6].

Nowdays, imaging modalities are always available to help in early diagnosis to identify the implantation site of an ectopic gestational sac. Once a woman with a missed period has an abnormally elevated β-hCG and no intrauterine pregnancy on transvaginal ultrasound, the diagnosis of an ectopic pregnancy can be established, even in the absence of histopathology of uterine curettage. Several cases have demonstrated that ultrasound combined with computed tomography (CT) plays a role in the early diagnosis of splenic ectopic pregnancy.

Ultrasound should be considered as the standard imaging tool of first choice. Screening pan-abdominal ultrasound can reveal a gestational sac-like echo image in the spleen, and color Doppler imaging can also show increased vascularity around the sac [3,7]. Rarely, embryos with a live fetal heart can be seen, as was in our case. Wu et al. published recently the first case of splenic pregnancy accurately diagnosed by ultrasound prior to treatment [8].

To confirm the results of an abdominal ultrasound, CT or MRI should be employed because they play an important role as diagnostic evidence and a detailed assessment of ectopic gestations, which helps to make management decisions. Although abdominal CT provided accurate diagnosis in most of previous case reports, it carries a risk of radiation exposure, therefore, MRI can be considered. Moreover, this method is increasingly available and useful in earlier or unruptured abdominal pregnancy. Our case was similar to that of Makrigiannakis et al, wherein both abdominal sonography and MRI verified the presence of an embryonic sac in the splenic parenchyma [5].

In unruptured splenic ectopic pregnancy cases, even if a patient with ruptured splenic pregnancy is hemodynamically stable or a non-surgical candidate, splenic preservation should be considered whenever possible because of its functional benefit. Several studies have been published showing successful conservative splenic treatment using minimally invasive approaches, and non-surgical management in combination with intramuscular methotrexate administration has been mentioned, including laparoscopic injection of methotrexate in the embryonic sac, CT-guided yolk sac aspiration with local injection of methotrexate, followed by ultrasound-guided percutaneous KCl injection, ultrasound-guided methotrexate injection [7], selective embolization of the splenic vessels feeding ectopic pregnancy by methotrexate [5] and partial splenectomy or splenorrhaphy [4] as well as our aforementioned case.

Conclusion

In reproductive-age females with abnormally elevated β-hCG levels and no intrauterine or pelvic pregnancy detected on

transvaginal ultrasound, it is advisable to examine patients using other imaging modalities, such as abdominal sonography, CT or MRI, to detect any upper abdominal pregnancies. In rare abdominal ectopic gestations, early successful diagnosis is essential because of the high risk of uncontrollable life-threatening intraperitoneal bleeding. Partial splenectomy is the treatment of choice with the benefit of splenic preservation, especially in unruptured splenic pregnancy.

Scientific Responsibility Statement
The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement
All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Conflict of interest
The authors declare no conflict of interest.

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